Plasma Cell Orificial Mucositis in a Pediatric Patient: Case Report

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INTRODUCTION

Plasma cell orificial mucositis (PCOM) is a very rare, benign plasma cell proliferative condition with an unknown etiology. It generally presents as erythematous mucosa with varying degrees of surface changes. It may be present on genitalia, lips, buccal mucosa, palate, gingiva, tongue, epiglottis, larynx, oropharynx, hypopharynx, and esophagus. PCOM can be asymptomatic, but depending on the location, may present with oral pain, dysphagia, dysgeusia, hoarseness, and/or sore throat, impacting quality of life. Currently, there are no associations with the development of a plasma cell neoplasm.

ETIOLOGY AND EPIDEMIOLOGY

- There are less than 50 cases currently reported, and only 2 cases previously reported in pediatric patients.
- Average age at the time of diagnosis is 56.6 years old.
- The cause is idiopathic, and the inheritance pattern is currently unknown.
- Most cases have been reported with a history of autoimmune or immunologically-mediated disease. However, no single disease has been found to be consistently associated. Previous cases have been reported with a history of Sjogren syndrome, autoimmune hepatitis, diabetes, and polymyositis.

DIAGNOSIS AND MANAGEMENT

- Diagnosis is based on health history, dental history, signs & symptoms, clinical presentation, and histological appearance. Histological analysis demonstrates the presence of dense polyclonal submucosal plasma cell infiltrate.

- Differential diagnosis includes: non-dental biofilm induced gingivitis, localized juvenile spongiotic gingival hyperplasia, Wegener's granulomatosis, mouth breathing, sarcoidosis, oral lichen planus, erythroplakia, oral squamous cell carcinoma, plasma cell gingivitis

- No standard treatment regimen has been established. Treatment regimens have limited success, short-term effectiveness, and may be ineffective. The goal of treatment is to reduce symptoms. Previously reported management options of PCOM involve:

  - Topical and/or systemic steroids
  - Immunosuppressive agents like methotrexate, tacrolimus, dapsone, mycophenolate mofetil, cyclosporine, colchicine, azathioprine infliximab, golimumab, adalimumab
  - Low-dose radiotherapy
  - Surgical intervention including resection, tracheostomy, laser excision
  - Frequent evaluation of soft tissue since tissue appearance of PCOM may mask malignancies or other gingival conditions
  - Spontaneous resolution

CASE REPORT

This case report presents a 10-year-old male patient who is a patient of the Children's Hospital Los Angeles. His medical history is significant for Fanconi anemia with a history of bone marrow transplant. He presented with erythematous tissue that had a strawberry-red appearance limited to the keratinized gingiva of the facial tissue of his maxillary and mandibular incisors (Figure 1). The patient reported bleeding upon brushing with no pain. The amount of plaque observed was not consistent with tissue changes present. A 2mm x 2mm x 2mm triangular tissue sample was collected from the keratinized tissue between teeth #7 and #8 to the level of the periosteum. The diagnosis of PCOM was made after histologic sections showed epithelial hyperplasia with spongiosis and a mixed subepithelial inflammatory infiltrate with numerous plasma cells and scattered neutrophils.

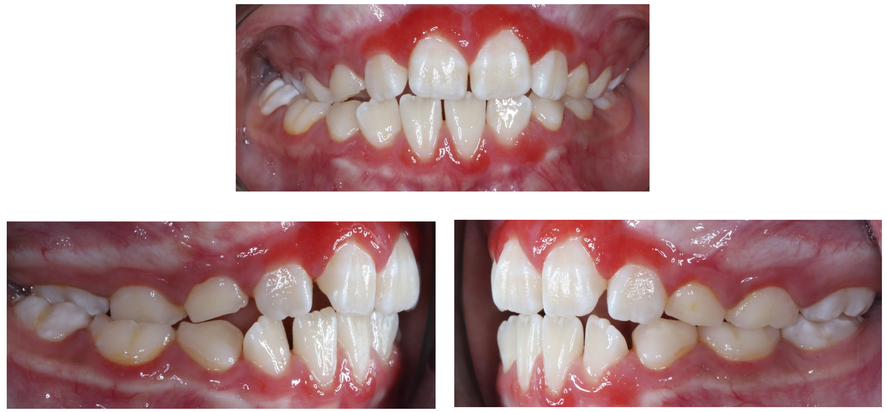

Figure 1. Frontal, Left and Right View. Tissue has a strawberry-red appearance limited to the keratinized gingiva of the facial tissue.

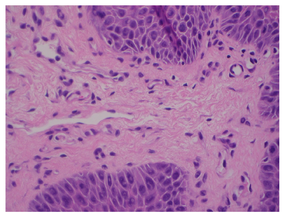

Figure 2. Normal tissue sample, not from patient. H&E, 40x: high power view of mucosa and gingiva with absence of pathology. Note the scarcity of lymphocytes and plasma cells.

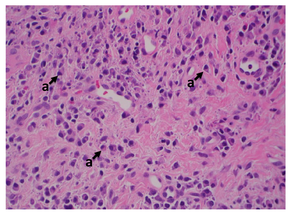

Figure 3. Tissue sample from patient. H&E, 40x: high power view of numerous plasma cells (a) demonstrating characteristic features: perinuclear clearings (hofs), eccentric nuclei, and occasional forms with clumped chromatin (clock face chromatin).

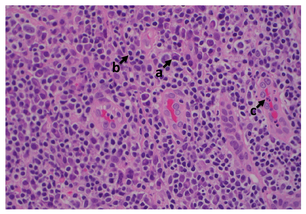

Figure 4. Tissue sample from patient. H&E, 40x: Histologic presentation of PCOM in a dense area of inflammation. Plasma cells (a) and lymphocytes (b) predominate within this field. Rare neutrophils (c) are seen coming out of the vessel on the right side.

DENTAL IMPLICATIONS and CLINICAL FEATURES

- The clinical presentation includes erythematous mucosa with varying degrees of surface changes.
- Excisional biopsy was conducted within lesion margins (Specimen: pink-tan, irregular mucosal tissue fragment measuring 0.5 x 0.4 x 0.1 cm)
- Histology demonstrated epithelial hyperplasia with spongiosis and a mixed inflammatory infiltrate (Figure 3) with numerous plasma cells and scattered neutrophils (Figure 4).

- Dental Implications:

  - Stringent oral hygiene to prevent increased inflammation of tissues.
  - Frequent recall visits to evaluate tissue health and changes.
  - Careful monitoring of tissue health during the course of dental and orthodontic treatment.
  - Collaboration with the medical team to determine appropriate treatment if the condition becomes symptomatic.

CONCLUSION

Plasma cell mucositis is a benign condition with a favorable prognosis that should be considered in the differential diagnosis of inflamed gingiva. Proper diagnosis of PCOM requires a biopsy and histological evaluation. If PCOM presents alongside autoimmune or immunologically-mediated diseases, it is important to collaborate with the patient's medical team to provide appropriate treatment and management. This case report presents the rare case of Plasma Cell Orificial Mucositis in a Pediatric Patient including its presentation and management.

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